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Sense of coherence in young people with congenital heart disease

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ABSTRACT

Objective: Patient-reported outcomes (PROs) have been found to play a role in the development of clinical complications. Hence, it is crucial to understand why some patients do well in terms of PROs and others do not and to identify these groups of patients. Sense of coherence (SOC), capturing a person's outlook on life, is associated with PROs in adolescents with congenital heart disease (CHD). Therefore, we (1) examine how SOC develops in young people with CHD, (2) identify subgroups of SOC development, and (3) characterize subgroups in terms of demographic and clinical variables and PROs.

Method: In this 4-wave longitudinal study, 429 adolescents with CHD (53.4% boys; median age = 16.3 years) completed assessments of SOC (SOC-13). PROs included quality of life (linear analog scale), loneliness (UCLA-8), depression (CES-D), and perceived health (PedsQL). Latent class growth analysis was used to identify clinically meaningful subgroups of SOC development.

Results: Patients with CHD had a moderate SOC that slightly decreased over the first 18 months. Four subgroups of SOC development emerged: *Consistently High* (27%), *Intermediate Stable* (41%), *Intermediate Decreasing* (25%), and *Chronically Low* (7%). Subgroups differed in terms of sex and PROs, but not in terms of age, disease complexity, primary diagnosis, or surgical history.

Conclusion: Patients with a strong and stable SOC over time showed a better adaptation than patients with a lower and/or decreasing SOC. Our results can guide the identification of patients at risk for adverse health outcomes and the development of interventions to enhance optimal living in patients with CHD.

Index terms: adolescence; heart defects, congenital; patient-reported outcomes; sense of coherence.

Survival in patients with congenital heart disease (CHD) has greatly improved.[1] As a result, health care professionals are increasingly interested in aspects of optimal living, such as quality of life (QOL), perceived health, and psychosocial functioning. These patient-reported outcomes (PROs) were found to play an independent role in the development of clinical complications in cardiac populations, such as adverse prognosis and hospitalization.[2] Hence, it is crucial to identify patients who demonstrate poor PROs both from the patient's perspective and from a clinical point of view.

In the literature, findings on PROs in youngsters with CHD are inconsistent.[3] On one hand, prior research has shown that adolescents with CHD may be psychosocially challenged, as evidenced by increased rates of psychological maladjustment, including internalizing (e.g., anxiety and depressive symptoms) and externalizing symptoms (e.g., aggression).[4, 5] Furthermore, recent findings indicated that youngsters with CHD score significantly lower on perceived health than controls.[6] On the other hand, recent studies found no differences between adolescents with and without CHD in terms of loneliness, anxiety levels, and emotional functioning.[7, 8, 9] One recent study by our group even showed that adolescents with CHD score better in terms of QOL as compared to controls.[10]

A meaningful psychological concept in relation to health outcomes is sense of coherence (SOC).[11] SOC is the core construct of the salutogenesis theory concentrating on the origins of health, in contrast to the pathogenesis theory focusing on the etiology of disease.[11] SOC represents a person's general outlook on life that characterizes the extent to which one appraises internal and external experiences and environments as comprehensible, manageable, and meaningful.[11] The theory of SOC explains why some individuals successfully adapt to major or chronic stressors, whereas others do not. Hence, one's ability to cope with stressors depends largely on the strength of one's SOC.[11] Generally, a strong SOC has been related to good QOL, physical, and psychosocial health.[12, 13] Furthermore,

SOC seems to be a highly actionable concept as its 3 components (i.e., comprehensibility, manageability, and meaningfulness) can inform interventions to improve SOC (e.g., educating patients to enhance comprehensibility).[14] SOC has been extensively studied in pediatric populations. Examples include attention-deficit hyperactivity disorder, asthma, autism, cerebral palsy, and epilepsy. Of note, findings concerning the influence of sex on adolescents' SOC are mixed, although a recent review concluded that in samples of older adolescents (≥15 years), males score higher on SOC as compared with females.[15]

Another well-known concept from the family of positive psychology is resilience, representing a person's capacity to rebound from negative experiences. As such, both resilience and SOC theories provide an answer to the question what makes people healthy despite being confronted with stressors. Because of the theoretical and empirical link between both concepts and the surrounding debate on possible overlap, a recent article by Fossion et al [16] focused on disentangling SOC and resilience. These researchers were able to identify a part of SOC that was independent from resilience and that significantly predicted depressive and anxiety disorders. The results of this study suggested that there is a part of SOC that is quite similar to resilience in that it mediated the relationship between undergoing multiple traumas and suffering from depression and anxiety and could change after negative experiences. Conversely, they argued that there is also a part of SOC that remains relatively stable and is not influenced by life events.[16] Hence, as previously suggested, SOC and resilience seem to be closely linked but distinct concepts. However, some studies use the SOC scale as a measure for resilience. In light of the aforementioned results, researchers should be careful in how they define and measure resilience.

Previous research in adolescents with CHD has shown that these patients have a higher mean SOC compared with age-matched controls.[10] In line with SOC theory, successfully managing and overcoming CHD-related problems can strengthen patients' SOC.[11, 14]

Conversely, the emerging SOC of an adolescent with CHD can also be weakened by severe health problems.[11] Furthermore, SOC has been associated with multiple PROs in adolescents with CHD. More specifically, SOC was negatively associated with anxiety and depression, and positively predicted QOL and perceived health over time.[5, 17, 18] Hence, research suggests that SOC may be of value in identifying patients at-risk for poor PROs who may benefit from specialized prevention or intervention programs. Nonetheless, important questions on SOC in young people with CHD remain largely unanswered. More specifically, although SOC mainly develops during adolescence and young adulthood, no longitudinal research has been published examining how SOC develops in young patients with CHD. Moreover, potential differences between patients in the development of SOC and the clinical value of such differences have not been investigated. Therefore, this study aims to categorize young people with CHD according to their SOC and has 3 objectives: (1) to examine the development of SOC during adolescence, (2) to identify distinct subgroups of individuals characterized by a similar development of SOC, and (3) to describe these subgroups in terms of demographic and clinical characteristics, and PROs.

METHODS

This study is part of the i-DETACH project (Information technology Devices and Education program for Transitioning Adolescents with Congenital Heart disease). In this longitudinal study, sense of coherence (SOC) was measured at 4 points in time with 9-month intervals in-between. Such relatively short time intervals were used to allow for a detailed mapping of development and change in this transitional stage. Demographic and clinical variables were measured at baseline (Time 1), and for this study, patient-reported outcomes (PROs) were examined after 27 months (Time 4).

Study Population and Procedure

Patients were selected from the database of the Department of Pediatric Cardiology and the Adult Congenital Heart Disease program at the University Hospitals Leuven (Belgium). Adolescents with congenital heart disease (CHD) could participate if they met the following criteria: aged 14-18 years at the start of the study in October 2009; confirmed CHD, defined as structural abnormalities of the heart and/or great intrathoracic vessels that are actually or potentially of functional significance [19]; last cardiac outpatient visit at the University Hospitals Leuven <5 years ago; Dutch-speaking; and valid contact details available. Patients were excluded if they met one of the following criteria: cognitive and/or physical limitations inhibiting filling out questionnaires, prior heart transplantation, and absence of informed consent. The targeted age range of 14-18 years means we only have a dropout of approximately 10%, as almost 90% of children with CHD reach the age of 14 years.[1] Of the 17,199 patients with CHD in the clinical database, a total of 498 patients met all inclusion criteria.

Eligible patients were sent a package by mail, containing an information letter, an informed consent form, a set of questionnaires, and a prestamped and addressed return envelope. Completing the entire set of questionnaires took approximately 60 minutes. Patients were asked to return the completed questionnaires within 3 weeks. To increase the response rate, adolescents received a movie ticket as an incentive for every measurement wave in which they participated. Furthermore, a modified Dillman's procedure was applied.[20] This procedure consisted of sending nonresponders a personalized reminder after 3 weeks, a formal reminder after 5 weeks, and contacting them by telephone after 7 weeks. Data were collected from the end of 2009 to the beginning of 2012. Written informed consent was obtained from all patients and/or their parents. The study protocol conforms to the ethical guidelines of the 1975 Declaration of Helsinki.

Measures

Sense of Coherence

Adolescents' SOC was measured using the 13-item Orientation to Life questionnaire.[11] A 7-point semantic differential scale ranging from 1 (very seldom or never) to 7 (very often) assessed the 3 components of SOC: comprehensibility (i.e., interpretation of the world as understandable, structured, and predictable; 5 items), manageability (i.e., the belief that resources are available to meet demands; 4 items), and meaningfulness (i.e., the belief that demands are challenges worthy of investment; 4 items). The total score ranges from 13 to 91 with a higher score indicating a stronger SOC. The validity and reliability of this scale are well established in the literature, also in adolescents with CHD.[21, 22] Cronbach's alpha coefficients for internal consistency ranged between .83 and .85.

Demographic and Clinical Characteristics

Information on patients' sex, age, disease complexity, primary diagnosis, and surgical history was obtained from chart review. Patients were identified as having a simple, moderate, or complex heart defect based on Task Force 1 of the 32nd Bethesda conference.[23] The primary heart defect was categorized using a modified version of the CONgenital CORvitia classification.[24]

Patient-Reported Outcomes

Four PRO domains were measured.

Quality of life

Based on previous conceptual work, quality of life (QOL) was defined as "the degree of overall life satisfaction that is positively or negatively influenced by individuals' perceptions of certain aspects of life important to them, including matters both related and unrelated to health".[25] Accordingly, QOL was rated by adolescents by marking a point on a vertically oriented, 10-cm line graded with indicators from 0 (worst imaginable QOL) to 100 (best

imaginable QOL). Previous research confirmed the validity and reliability of this measure in patients with CHD.[25]

Loneliness

The 8-item version of the University of California Los Angeles Loneliness Scale (UCLA-8) was used. Adolescents responded to each item using a scale ranging from 1 (strongly disagree) to 5 (strongly agree). A scale score was calculated by averaging all the items and, thus, a higher score reflected higher levels of loneliness. This scale proved to be valid and reliable for use in healthy adolescents,[26] and internal consistency proved to be good in adolescents with CHD.[7] Cronbach's alpha ranged between .81 and .86.

Depressive symptoms

Depressive symptoms were measured with the 20-item Center for Epidemiologic Studies Depression Scale (CES-D). Each item measured the frequency at which participants experienced symptoms of depression during the past week, using a scale ranging from 0 (seldom) to 3 (most of the time or always). The total score ranges from 0 to 60 with higher scores indicating more frequent depressive symptoms. The psychometric properties of this scale have been confirmed.[27] Cronbach's alphas ranged between .74 and .76.

Perceived health

Domains of perceived health were measured with the Pediatric Quality of Life Inventory 4.0 (PedsQL; Mapi Research Trust, Lyon, France). The PedsQL was originally developed to measure health-related QOL. However, this does not fully fit with how QOL was defined in this study. Indeed, researchers often use the term QOL when actually health status might be a more suitable term.[28] Therefore, the PedsQL more likely expresses domains of perceived health. Four domains of generic perceived health were measured: physical, emotional, social, and school functioning. Five domains of disease-specific perceived health were included: symptoms, physical appearance, treatment anxiety, cognitive

problems, and communication. The degree to which adolescents experienced problems during the past month was rated on a scale ranging from 0 (never) to 4 (almost always). These scores were then transformed into a 0 to 100 scale with a higher score reflecting better perceived health. The PedsQL scale has proven to be reliable and valid.[29] Cronbach's alphas ranged between .89 and .90 for generic perceived health and between .88 and .90 for disease-specific perceived health.

Statistical Analysis

Statistical analysis comprised 2 parts. First, to examine how SOC develops over time, Latent Class Growth Analysis (LCGA) was conducted in Mplus 5.0 (Muthén & Muthén, Los Angeles, CA). LCGA is a group-based, semiparametric approach based on finite mixture modeling.[30] More specifically, LCGA is a person-centered approach that summarizes longitudinal data by classifying individuals into distinct subgroups or classes, each group consisting of individuals who show a similar development of SOC over time. Identified subgroups are characterized by unique initial levels (intercepts) and rates of change (slopes). To identify the optimal number of subgroups within our data, LCGA models for 1 to 6 classes were run. For each model, the path from the slope to the indicator at Time 1 was fixed to zero so that the intercept represented the initial level. Given the equally spaced measurement intervals of 9 months, linear slope pattern coefficients were fixed at 0, 1, 2, and 3 for Times 1, 2, 3, and 4, respectively. Several fit indices were used to decide on the number of subgroups.[30] Firstly, the Bayesian Information Criterion (BIC) statistic for a solution with *k* classes should be lower than the BIC value for a solution with *k*-1 classes, suggesting that adding classes improves the model fit. Furthermore, entropy values of ≥ 0.75 indicate accurate classification of individuals into subgroups based upon posterior probabilities of classification.[31] Finally, the number of patients per subgroup should represent more than 5% of the sample.

Second, to describe the subgroups obtained through LCGA, χ^2 tests, Kruskal-Wallis *H* tests, and multivariate analyses of variance with pairwise comparisons using Gabriel's test procedure (i.e., post hoc tests) were conducted. These analyses were conducted in SPSS version 20.0 (SPSS Inc., Chicago, IL) with the significance level at $p \leq .05$ (2-tailed).

RESULTS

Sample Characteristics

In total, 429 adolescents with congenital heart disease (CHD) (86%) participated at Time 1, 398 participated at Time 2, 366 participated at Time 3, and 337 participated at Time 4. Overall, 304 (71%) patients participated at all subsequent measurements. Participants with and without complete data were compared using Little's missing completely at random test, which revealed a normed chi-square (χ^2/df) of 1.06, indicating that all missing values could be reliably estimated.[32] Accordingly, to deal with missing values, we used the expectation-maximization algorithm. As such, all analyses were performed on the full sample of 429 participants. As detailed in Table 1, the sample consisted of 229 boys and 200 girls with a median age of 16.3 years (interquartile range 1.9). The majority of these patients had a moderate defect (47.6%) and the most frequent primary diagnosis was a ventricular septal defect (18.1%). In total, 46.6% of these patients underwent prior heart surgery (Table 1). Other demographic and clinical sample characteristics are reported elsewhere.[17]

Table 1. Patient characteristics for the total group and the 4 sense of coherence subgroups

		Sense of coherence subgroup				
Variable	**Total group (n=429)**	**Consistently High (n=115)**	**Intermediate Stable (n=174)**	**Intermediate Decreasing (n=108)**	**Chronically Low (n=32)**	***p***
Median age at Time 1[a]	16.3 (1.9)	16.3 (1.8)	16.3 (2.8)	16.1 (1.8)	16.6 (1.8)	0.673[b]
Sex, %[c]						0.004[d]
Boys	53.4	67.0	50.6	48.1	37.5	
Girls	46.6	33.0	49.4	51.9	62.5	
Disease complexity, %[c]						0.378[b]
Simple	40.6	41.7	43.1	33.3	46.9	
Moderate	47.6	48.7	44.8	52.8	40.6	
Complex	11.9	9.6	12.1	13.9	12.5	
Top 5 primary heart defect diagnoses (n=284), %[c]						0.790[d]
Ventricular septal defect	18.1	18.3	20.7	14.8	15.6	

Aortic valve abnormality	16.0	19.1	16.7	13.0	12.5	
Atrial septal defect type II	13.1	11.3	12.1	15.7	15.6	
Coarctation of the aorta	10.0	10.4	9.8	10.2	9.4	
Pulmonary valve abnormality	8.9	10.4	5.7	10.2	15.6	
Surgical history before Time 1, %[c]						0.282[d]
Yes	46.6	44.3	46.6	52.8	34.4	
No	53.4	55.7	53.4	47.2	65.6	

[a]Interquartile ranges (medians) are mentioned in parentheses. [b]Kruskal-Wallis *H* test. [c]Values indicate the percentages within sense of coherence subgroups (i.e., within columns). [d]Chi-square test.

Development of Sense of Coherence

Looking at the 1-class solution of the Latent Class Growth Analysis (LCGA), moderate levels of sense of coherence (SOC) were found over time in the total sample (intercept = 4.73; $p < .001$). Further, there was a slight decrease in SOC across the first 3 measurements, as shown by a significant negative linear slope (-0.09; $p < .01$) and positive quadratic slope (0.02; $p < .05$). LCGAs favored a 4-class solution over a 3-class solution as evidenced by a lower Bayesian Information Criterion (BIC) value (BIC = 3530.27 vs BIC = 3598.25). Additionally, the entropy value of the 4-class solution (0.84) suggested good classification accuracy. Furthermore, the 4-class solution was more parsimonious as compared with the 5-class solution, in which one of the classes represented a variation on one single theme and consisted of only 5% of the sample. Hence, the model with 4 classes was selected. Fit indices, subgroup frequencies, intercepts, and both linear and quadratic slope terms associated with these models are shown in Table 2. As the 6-class solution had a higher BIC value than the 5-class solution (i.e., 3518.67), this solution is not represented in Table 2.

A graphical presentation of the observed mean SOC values for the 4 subgroups of SOC development is provided in Figure 1. In general, the subgroups of SOC development differed mainly in terms of the level of SOC and not so much in terms of changes in SOC over time. Class 1 (27% of the sample) was labeled *Consistently High* and was characterized by persistently high scores on SOC. Class 2 represented the majority of our sample (41%), with moderate SOC scores over time remaining stable (*Intermediate Stable*). The third class (25%) consisted of patients who demonstrated moderate levels of SOC at baseline. In contrast to Class 2, these patients' SOC decreased especially across the first 3 time points (*Intermediate Decreasing*), as shown by the significant negative linear slope and positive quadratic slope (Figure 1). Finally, Class 4 (7%) consisted of patients with a consistently low SOC over time (*Chronically Low*).

Table 2. Results of different latent class growth analyses in adolescents with congenital heart disease

Solution	BIC	Entropy	Sense of coherence subgroup				
			1	2	3	4	5
1-class	4547.14	/					
Proportion			429				
Mean intercept			4.73***				
Mean linear slope			-0.09**				
Mean quadratic slope			0.02*				
2-class	3872.13	0.81					
Proportion			198 (46%)	231 (54%)			
Mean intercept			4.10***	5.28***			
Mean linear slope			-0.18***	-0.02			
Mean quadratic slope			0.05**	0.003			
3-class	3598.25	0.84					
Proportion			192 (45%)	103 (24%)	134 (31%)		
Mean intercept			4.68***	3.76***	5.57***		

Mean linear slope			-0.12*	-0.21**	0.02		
Mean quadratic slope			0.03†	0.05**	-0.01		
4-class	**3530.27**	**0.84**					
Proportion			**108 (25%)**	**32 (7%)**	**115 (27%)**	**174 (41%)**	
Mean intercept			**4.14*** **	**3.15*** **	**5.62*** **	**4.79*** **	
Mean linear slope			**-0.26*** **	**-0.12**	**0.04**	**-0.08**	
Mean quadratic slope			**0.06****	**0.05**	**-0.02**	**0.02**	
5-class	3513.51	0.83					
Proportion			30 (7%)	116 (27%)	20 (5%)	104 (24%)	159 (37%)
Mean intercept			3.13***	5.44***	6.06***	4.11***	4.74***
Mean linear slope			-0.12	0.01	0.13	-0.27***	-0.09
Mean quadratic slope			0.05	-0.004	-0.05	0.06**	0.02

The selected solution is represented in bold. The BIC for the 6-class solution (3518.67) was higher than for the 5-class solution and, hence, the 6-class solution is not represented here. *$p < .05$; **$p < .01$; ***$p < .001$; †$p < .10$. BIC, Bayesian Information Criterion.

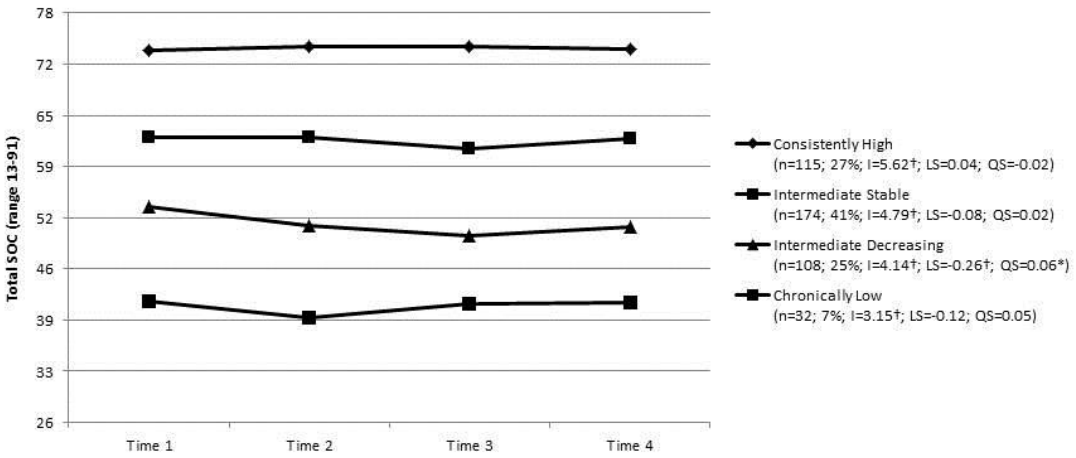

Figure 1. Subgroups of sense of coherence development. Observed mean values of sense of coherence (SOC) and predicted subgroups in development for young patients with congenital heart disease across 4 time points. Proportions of patients, mean intercepts (I), mean linear slopes (LS), and mean quadratic slopes (QS) are in parentheses. *$p < .01$. †$p < .001$.

Characteristics of the Subgroups of Sense of Coherence Development

Table 1 describes baseline demographic and clinical characteristics for the 4 subgroups of SOC development. Chi-square analysis showed a significant association between patients' sex and subgroup allocation ($\chi^2(3) = 13.50$, $p = .004$). More boys belonged to the *Consistently High* and *Intermediate Stable* subgroups as compared with girls. Conversely, more girls belonged to the *Intermediate Decreasing* and *Chronically Low* subgroups as compared with boys (Table 1). No significant differences were found with regard to age, disease complexity, the 5 most frequent primary diagnoses, or surgical history (Table 1).

In terms of PROs, Table 3 shows the mean values of QOL, loneliness, depressive symptoms, and perceived health for all subgroups and differences among these subgroups, after controlling for patients' sex and disease complexity. All subgroups differed significantly from each other in terms of QOL, loneliness, and depressive symptoms. As a general pattern, scores on QOL, loneliness, and depressive symptoms worsened with lower SOC scores. As such, young people from the *Consistently High* subgroup reported the highest levels of QOL and the lowest scores on loneliness and depressive symptoms. In contrast, patients from the *Chronically Low* subgroup scored lowest on QOL and highest on loneliness and depressive symptoms (Table 3).

A similar pattern of results emerged regarding generic and disease-specific domains of perceived health, with the highest scores on domains of perceived health in the *Consistently High* subgroup and the lowest scores in the *Chronically Low* subgroup. All subgroups differed significantly from each other for the domains of emotional and social functioning, symptoms, cognitive problems, and communication. Furthermore, for physical functioning, the *Intermediate Decreasing* and *Chronically Low* subgroups differed significantly from each other, and from the *Consistently High* and *Intermediate Stable* subgroups. For school functioning and physical appearance, the *Consistently High* and *Intermediate Stable*

subgroups differed significantly from each other and from the *Intermediate Decreasing* and *Chronically Low* subgroups. Finally, for treatment anxiety, the *Consistently High* subgroup differed significantly from the *Intermediate Decreasing* and *Chronically Low* subgroups, and the *Intermediate Stable* subgroup also scored significantly better than the *Chronically Low* subgroup (Table 3).

Table 3. Analyses of variance and post hoc comparisons based upon Gabriel's pairwise test procedure for the 4 sense of coherence subgroups

	Sense of coherence subgroup: Mean (SD)						
Variable	**Consistently High (n=115)**	**Intermediate Stable (n=174)**	**Intermediate Decreasing (n=108)**	**Chronically Low (n=32)**	**λ**	***F* value**	**η^2**
Time 4					0.54	7.49*	0.19
Quality of life	86.54 (7.43)[a]	82.47 (9.14)[b]	77.30 (9.44)[c]	68.98 (14.59)[d]		25.20*	0.16
Loneliness	1.44 (0.38)[a]	1.75 (0.52)[b]	2.19 (0.65)[c]	2.64 (0.79)[d]		30.11*	0.18
Depressive symptoms	4.57 (3.30)[a]	9.08 (6.25)[b]	15.68 (7.38)[c]	24.31 (10.57)[d]		58.80*	0.30
Generic domains of perceived health							
Physical functioning	90.87 (10.53)[a]	88.23 (11.73)[a]	82.23 (12.76)[b]	72.15 (15.52)[c]		15.85*	0.11
Emotional functioning	84.50 (11.94)[a]	75.88 (13.18)[b]	63.79 (14.74)[c]	49.01 (15.08)[d]		52.60*	0.28
Social functioning	91.48 (10.19)[a]	87.54 (11.87)[b]	80.77 (13.35)[c]	69.98 (17.74)[d]		18.00*	0.12
School functioning	86.01 (11.18)[a]	79.31 (12.18)[b]	69.22 (13.47)[c]	63.09 (16.50)[c]		28.77*	0.18
Disease-specific domains of perceived health							
Symptoms	85.92 (12.49)[a]	81.01 (14.95)[b]	74.06 (16.23)[c]	61.80 (18.46)[d]		12.88*	0.09

Physical appearance	88.32 (12.10)[a]	81.17 (16.10)[b]	67.81 (21.56)[c]	62.07 (17.29)[c]	22.38*	0.14
Treatment anxiety	90.10 (15.37)[a]	86.06 (17.87)[a,b]	80.89 (20.55)[b,c]	72.75 (27.36)[c]	11.16*	0.08
Cognitive problems	83.03 (15.70)[a]	75.62 (16.46)[b]	65.16 (16.95)[c]	53.72 (20.78)[d]	24.91*	0.16
Communication	88.75 (13.63)[a]	82.31 (16.58)[b]	72.13 (19.71)[c]	56.11 (21.94)[d]	26.60*	0.17

Multivariate differences (Wilks' Lambda, λ), follow-up univariate *F* values and effect sizes (η^2) are reported. A subgroup mean is significantly different from another mean if they have different superscripts (i.e., a, b, c, and d). For example, for treatment anxiety, the *Consistently High* and *Intermediate Stable* subgroups share the same superscript (i.e., a) and, thus, are not significantly different from each other. Conversely, the *Consistently High* subgroup has a different superscript than the *Intermediate Decreasing* subgroup (i.e., a vs b, c) and, thus, these subgroups differ significantly from each other in terms of treatment anxiety. Standard deviations (SD) are put in parentheses. *$p < .001$.

DISCUSSION

In the total sample of young people with congenital heart disease (CHD) sense of coherence (SOC) scores were moderate and slightly decreased over the first 18 months. Latent Class Growth Analysis (LCGA) identified 4 substantially different subgroups of SOC development: (1) *Consistently High*, (2) *Intermediate Stable*, (3) *Intermediate Decreasing*, and (4) *Chronically Low*. The small, decreasing trend in the development of SOC was only present in the *Intermediate Decreasing* subgroup as SOC remained relatively stable among all other subgroups. This general tendency towards stability of SOC seems to suggest that frequent short-term measurements of SOC during adolescence are unnecessary in future research.

From a clinical perspective, it is crucial to identify patients who are on a pathway to maladjustment and ill-being, thereby allowing timely intervention. As such, the findings of this study have several important clinical implications. First, although previous research found that SOC in adolescents with CHD was significantly higher as compared with healthy peers,[10] our results show that a subgroup of patients deteriorate over time regarding their resistance to stress (i.e., *Intermediate Decreasing* subgroup). Moreover, a small subset of patients has a persistently low SOC over the 4 measurement waves (i.e., *Chronically Low* subgroup). These 2 groups accounted for almost one-third of the participants (32%). Hence, this longitudinal study shows that LCGA could help in distinguishing patients who do well in terms of SOC from patients who struggle.

Second, girls tended to have a lower SOC, which is consistent with previous research in adolescents with and without CHD.[15, 33] This could be because girls experience higher levels of interpersonal stress and more negative life events than their male peers.[34, 35] The fact that girls appear to perceive life as less comprehensible, manageable, and meaningful might render them more susceptible to negative health development. However, the 4

subgroups did not differ from each other in terms of age, disease complexity, primary diagnosis, and surgical history. Previous research also concluded that the cardiac diagnosis did not relate to psychosocial functioning in young patients with CHD.[36] Hence, health care professionals should pay attention to patients with all types of CHD.

Third, scores on patient-reported outcomes (PROs) worsened with lower SOC scores. In general, all 4 subgroups were clearly distinguishable in terms of QOL, loneliness, depressive symptoms, and domains of perceived health. The fact that patients with a chronically low SOC suffered most from depressive symptoms, loneliness, lower QOL, and lower perceived health highlights the negative effects associated with a low SOC. Hence, this study identified a potential pathway for targeting at-risk youth as psychosocial variables have been associated with adverse clinical outcomes in other cardiac populations.[2] This most vulnerable group of young people with CHD could benefit from a timely referral to psychosocial care addressing their needs and preventing further deterioration of their health and well-being. For example, health care professionals could focus on enhancing patients' SOC through lifestyle interventions.[37] Furthermore, in daily clinical practice, health care professionals could anchor medical encounters around the 3 actionable components of SOC by providing information (comprehensibility), by making patients aware of available resources (manageability), and by supporting patients to participate actively in their care (meaningfulness).[14]

This study had a number of limitations. First, our data are limited to self-reports, which may lead to single-reporter bias. Logically, PROs are best captured by self-reported instruments as patients' views are not necessarily the same as their (adult) proxies'. However, studies on patients with CHD have previously highlighted the importance of collecting data from different sources to prevent bias due to denial or underestimation of problems.[38] Second, the period over which patients were followed was relatively short, which is probably

the reason why we found little change in the development of SOC. Although Antonovsky argued that SOC mainly develops during the first 3 decades of a person's life, researchers found that SOC may increase over time well after the third decade of life.[39] However, studies also found that people who start off with a strong SOC show greater stability in their SOC throughout adulthood.[11, 39] In sum, referring to the state-trait distinction upheld in various domains of personality research, one could argue that there may be a trait aspect of SOC that shows only limited changes throughout the life span after it is established in the first decades of life. Other, more state-like, aspects of SOC might be more amenable to change and might show more changes after certain life challenges and stressors.[16] Long-term longitudinal research with a continuous follow-up spanning the entire range of adolescence and adulthood would provide additional information. Third, significant differences among subgroups of SOC development provide a first insight in potentially relevant factors for clinical practice but must not be interpreted in terms of causality. To examine the predictive value of SOC or to identify predictors of SOC within a longitudinal data set, cross-lagged path analysis could be applied. Fourth, the generalizability of our results is limited because of the single-center setting and the specific nature of our sample.

Some of these limitations can be seen as fruitful avenues for future research. First, continuing longitudinal research is needed to evaluate whether our findings extend into adulthood. Furthermore, multicentric international research is required examining the generalizability of previous results concerning SOC and its relation with health and well-being in patients with CHD. Such a cross-cultural perspective is an essential step in defining the importance and utility of SOC. In addition, future research would benefit from including more information on patients' medical condition (e.g., functional capacity) to establish the relationship between the development of SOC and clinical outcomes more firmly. Finally,

intervention studies should investigate the possibilities of improving patients' health and well-being.

CONCLUSIONS

Substantial individual differences in the development of sense of coherence (SOC) were observed as 4 subgroups of SOC development were identified. These subgroups were meaningfully differentiated in terms of patient-reported outcomes in young people with congenital heart disease (CHD). More specifically, patients with a strong and stable SOC over time showed a better adaptation than patients with a lower and/or decreasing SOC. As such, these results outline a potential method for identifying young people with CHD suffering from greater psychosocial struggles and who, due to their cardiac history, may be at greater risk for adverse clinical outcomes. Hence, stratifying patients according to their SOC can help initiating early psychosocial interventions enhancing optimal living in patients with CHD.

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